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Acantholytic squamous cell carcinoma of oral cavity, mimicker of angiosarcoma: A rare case report

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ABSTRACT

Acantholytic squamous cell carcinoma (ASCC) is an uncommon variant of SCC. It was first described by Lever in 1947. It usually develops on sun-exposed areas of older-aged individuals. Histologically, the tumor consists of a nodular, epidermal-derived proliferation that forms islands like structures. At least focally or sometimes extensively, the tumor cells show a loss of cell cohesion within the central gland-like or tubular spaces. The lesions present as slowly growing scaly and occasionally ulcerated papules or plaques. Areas of acantholysis may produce large intraepidermal cavities which may extend to the adjacent follicular structures. ASCC is rarely seen in the oral cavity. Here, we report an unusual case of ASCC of the oral cavity, which was initially misinterpreted as angiosarcoma on histopathology. Then, on immunohistochemistry, ASCC was confirmed.

Key words: *Acantholytic, Acantholytic squamous cell carcinoma, Angiosarcoma, Squamous cell carcinoma*

Acantholytic squamous cell carcinoma (ASCC) is an uncommon variant of SCC characterized by a loosening of the intercellular bridges, resulting in acantholysis. These tumors may present as intraepidermal (*in situ*) or invasive SCC [1]. There is a formation of anastomosing spaces and channels mimicking an angiosarcoma [2]. The overall incidence of ASCC is only 0.1%, and ASCC by itself is a rare entity at any site in the body. ASCC in the oral cavity is extremely rare. The peak incidence of oral ASCC is about sixth or seventh decade [3]. ASCC is known to cause diagnostic difficulties to the pathologist due to the discohesive pattern it generates. Hence, there is a multitude of terminologies associated with the entity such as adenoid SCC, pseudoglandular SCC, and angiosarcoma-like SCC [4]. Here, we report a rare case of ASCC of the oral cavity in a 45-year-old male.

CASE REPORT

A 45-year-old male presented with complaints of swelling and pain in the right gingival region for 6 months. The pain was intermittent in nature and aggravated on mastication and relieved itself after few minutes. He was a chronic smoker and gutkha chewer for 15 years. He was eating 4–5 packets of gutkha and smoking 3–4 cigarettes daily.

On local examination, a well-defined brownish nodular growth was present in the gingivobuccal sulcus in the region of 1st and 2nd molar teeth. The surface of the nodule was eroded at places, soft in consistency, brown in color, and non-indurated. It was bleeding on touch (Fig. 1). There was a large area of leukoplakia on the right buccal mucosa posterior and lateral to the growth. Radiograph examination was done and showed no bony involvement. Clinical diagnosis of pyogenic granulomas was made, and the lesion was excised. Tumor mass was adherent to the teeth, so extraction of the teeth was also done during excision of the tumor. Post-operatively after 10 days, the patient came for follow-up with a large growth in the same region.

Histopathology revealed large ovoid loose cells with moderate amount of cytoplasm and high nucleocytoplasmic ratio, high vascularity, and high mitotic activity (Figs. 2 and 3). Histopathology report was given as intermediate grade angiosarcoma. Immunohistochemistry was advised for confirmation of the diagnosis. Immunohistochemistry revealed tumor cells positive for cytokeratins (CK) (AE1/AE3), CK5/6, and CK8/8 (focal) and immunonegative for epithelial membrane antigen (EMA), CD31, CD34, and ERG. The final diagnosis of ASCC was offered. Then, he was referred for radical surgery to the higher center. Right hemimandibulectomy with radical neck dissection was done. Post-operatively chemotherapy and radiotherapy were advised; however, the patient lost to follow-up after the radical surgery.

DISCUSSION

ASCC is a well-recognized, uncommon variant of SCC. It was first described by Lever in 1947. It usually develops on the sun-exposed skin, predominantly in the head and neck region, in particular on and around the ears. It is rarely seen on the mucosal surface of an upper aerodigestive tract [5]. The World

Figure 1: Clinical photograph showing well-circumscribed, soft, brownish nodular growth in gingivobuccal sulcus

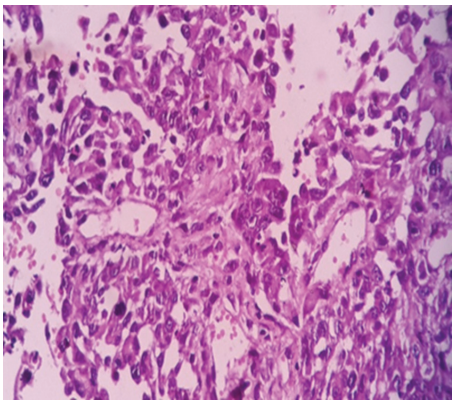

Figure 2: Photomicrograph showing loosely arranged cells with increased vascularity, mitosis, and pseudoglandular spaces (H and E stain, ×400)

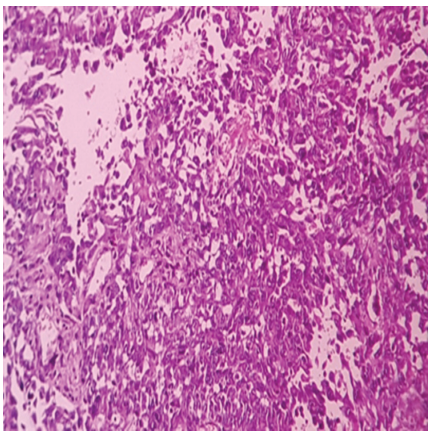

Figure 3: Photomicrograph showing dissociated cells with cystic spaces and increased vascularity (Hand E stain, ×400)

Health Organization defined ASCC as an original entity since a long time, but there are <30 cases of ASCC documented in the international literature so far [3].

Histologically, ASCC and angiosarcoma are completely different tumors. In our case, many cystic and vascular spaces appear to be lined by tumor cells. The tumor cells were dissociated and loosely arranged. There was a high degree of pleomorphism, anisonucleosis, and many atypical mitoses. Considering these features, histopathologically misinterpreted as angiosarcoma. Although angiosarcoma and ASCC are completely different tumors, their histological features are similar and defined by intratumoral spaces. Formation of anastomosing spaces and channels in ASCC closely resembles angiosarcoma. Both tumor entities show comparable clinical appearance in the oral cavity. Macroscopically both entities located in the oral cavity are fast growing, eruptive lesions, and have a poor prognosis [6,7].

The age range of ASCC is wide, but it usually affects aged individuals with male predominance. Angiosarcoma is also common in elderly males. Clinically, ASCC presents as a circumscribed white, gray ulcer, or nodular tan red/pink tumor, often located on the sun-exposed areas. Angiosarcoma begins as a very poorly defined red plaque resembling a bruise. Lesions can become quite large, and areas of nodularity may arise. In ASCC, epithelial markers such as CK and EMA are positive, and these are negative for CD31 and factor VIII-related antigens. In angiosarcoma, epithelial markers are negative and vascular markers are positive. Prognosis of angiosarcoma is poor as compared to ASCC [1,2].

ASCC is a master mimic on histopathology. It is known to cause diagnostic difficulties to the pathologist due to discohesive pattern it generates. Hence, there is a multitude of terminologies associated with this entity such as adenoid SCC, pseudoglandular SCC, SCC with gland-like features, angiosarcoma-like SCC [8], and pseudovascular adenoid SCC [9]. ASCC differs from common SCC not only histologically but also by its aggressive behavior. When it occurs in the oral cavity, it is associated with a poor prognosis [10].

CONCLUSION

As the overall incidence of this tumor entity is very rare, the lack of understanding of this tumor poses diagnostic dilemma to the pathologist. Meticulous observation of the morphological pattern is important for the diagnosis. It should be differentiated from angiosarcoma and adenosquamous carcinoma.

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